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Review article

# Hyperlexia: Systematic review, neurocognitive modelling, and outcome



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#### ABSTRACT

Hyperlexia is defined as the co-occurrence of advanced reading skills relative to comprehension skills or general intelligence, the early acquisition of reading skills without explicit teaching, and a strong orientation toward written material, generally in the context of a neurodevelopmental disorder. In this systematic review of cases (N=82) and group studies (including 912 participants of which 315 are hyperlexic), we address: whether the hyperlexic profile is associated with autism and why, whether models of non-autistic reading can teach us about hyperlexia, and what additional information we can get from models specific to autistic cognitive functioning. We find that hyperlexia, or a hyperlexic-like profile, characterises a substantial portion of the autistic spectrum, in which the subcomponents of the typical reading architecture are altered and dissociated. Autistic children follow a chronologically inverted path when learning to read, and make extended use of the perceptual expertise system, specifically the visual word form recognition systems. We conclude by discussing the possible use of hyperlexic skills in intervention.

#### 1. Introduction and definition

The term "hyperlexia" was coined in 1967 by Silberberg and Silberberg (1968), Silberberg and Silberberg (1967). Four features consistently describe hyperlexia: (1) the presence of an accompanying neurodevelopmental disorder; (2) advanced reading skills, relative to comprehension skills or general intelligence; (3) an early acquisition of reading skills without explicit teaching; and (4) a strong orientation toward reading material (Needleman, 1982). The same year, Rutter and Lockyer noticed that the same proportion of both their groups of "psychotic children" and control children could read at a normal level for their age, despite very little schooling for the "psychotic" group. They described the example of a boy who could read well above the average level, although he had been excluded from school at the age of six (Rutter and Lockver, 1967). However, cases of hyperlexia were described as early as 1930 (Phillips), i.e. before the introduction of both hyperlexia and autism as a syndrome. Mentions of the hyperlexic profile can be found even earlier, in 1918, when Hollingworth and Winford wrote, "Cases where a generally stupid (sic) child is innately gifted with special ability to master the mechanics of reading, for

example, are no doubt as frequent as cases where a generally capable child learns them with difficulty." (Hollingworth and Winford, 1918; cited in Thompson, 1966; p.24).

Fifty years after the term was first introduced, hyperlexia is often reported as one of the savant abilities in autism. However, its neurocognitive underpinning and how it relates to autistic cognition and typical reading acquisition, are yet to be established. Literature on hyperlexia consists of a large number of descriptive reports, combined with a small number of empirical studies testing specific assumptions about its prevalence and mechanisms. Beyond the established cross-sectional differences in decoding and understanding abilities, the distinct, developmental pathway of hyperlexics remains to be characterized. The last academic review on hyperlexia was published 14 years ago (Grigorenko et al., 2003). Multiple case and group studies have been published in the last decade, including the first imaging studies of hyperlexia, making a new review necessary.

Based on a systematic review, we first report the current state of knowledge on the prevalence of hyperlexia and its relation with autism. We present the cognitive processes underlying reading in non-hyperlexic, mature readers, and the sequence of typical reading acquisition.

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Table 1
Systematic review of hyperlexic case reports – \*The number of criteria for hyperlexia refers to the four-feature definition given in section 1. \*\*The oral language level is classified based on the indications used to determine the ADOS module: 1 refers to individuals with no speech, or who do not consistently use phrase speech; 2 refers to individuals with some phrase speech (defined as non-echoed three-word utterances that sometimes involve a verb and are spontaneous, meaningful word combinations), but not verbally fluent; 3 refers to fluent speech. Verbal fluency is broadly defined as having the expressive language of a typical four-year-old child: producing a range of sentence types and grammatical forms, using language to provide information about events, and producing logical connections within sentences (e.g., "but" or "though"). \*\*\*The autistic diagnosis is determined as explained in 3.2. Additional information on the scoring system can be obtained from the authors on request.

Millic	Publication	Case	Gender	Age at time of report (y = years, m = months)	Hyperlexic features mentionned <sup>*</sup> (1–4)	Reported onset of hyperlexia	Oral language level <sup>**</sup>	Autism-PDD mentionned	Autistic traits based on description***	Other condition mentionned- not autistic	Not enough information for autism diagnosis
Care											
Silberberg   Case 2   M				•		·					
Case 3	Silberberg and	Case 1	F	7y1m	4	NA	2				X
Case 4   M   Grade 2   M   NA   1	Silberberg	Case 2	M	Grade 4	4	NA	2				X
Case 5	(1968)	Case 3	F	Зу	4	18 months	2	X			
Case 1		Case 4	M	Grade 2	4	NA					
Cain (1969)   Case 1:   F   695m   4   NA   1   X   NA   1   X   NA   1   X   NA   1   X   NA   X											
Mille		Case 6	M	Grade 3	4	NA	3				X
Case 2:	Cain (1969)		F	6y5m	4	NA	1	X			
Case 3:		Case 2:	F	7y	4	NA	2	X			
Mehegan and   Case   1 6 6.0   M   6y   4   NA   1   X		Case 3:	M	8y	4	NA	1	X			
Mehegan and   Case   1 6 6.0   M   6y   4   NA   1   X	Goodman (1972)	Case: Sam	M	7v11m	4	Before 4v	2	x			
Definis   Case 2				-		•					
Case	Dreifuss										
Huttenlocher and Case 1: M.K. M. 95						-					
Huterlocher   Case 2: C.O. M		Case 4: 8.0			3	-	1	X			
Cobrinik, 1974  Case 1: M			M								
Cobrinik, 1974			M	4y11m							
Anthony B.  Case 2:  M 14y6m 4 Before 5y 2 X  Robert C.  Case 3:  M 14y 4 Before 5y 2 X  Terence D.  Case 4: John M 15y 4 Before 5y 1 X  M.  Case 5:  M Malcolm P.  Case 6:  William W.  Case 6:  William W.  Case 6:  William W.  Case 1: V. F 5y8m 3 1y3m 1 X  Selection of 1976)  Aram et al. (1984) Case: MD M 39y 4 4y5m 2 X  Gase 1: M.H. F 17y8m 4 3y 2 X  Case 2: D.M. M 6y8m 4 3y 2 X  Case 2: D.M. M 6y8m 4 3y 2 X  Case 4: D.S. M 5y3m 4 Before 4y 1 X  Case 4: D.S. M 5y3m 4 Before 4y 1 X  Case 6: C.R. M 10y1m 4 2y 2 X  Case 6: T.M. M 12y6m 3 4y6m 2 X  Case 6: T.M. M 12y6m 3 2y 2 X  Soldberg and Rothermel (2se 7: L.N. M 5y2m 4 3y 2 X  Case 6: T.M. M 12y6m 3 4y6m 2 X  Case 6: T.M. M 12y6m 3 2y 2 X  Soldberg and Rothermel (2se 7: L.N. M 5y2m 4 3y 2 X  Case 6: T.M. M 12y6m 3 4y6m 2 X  Case 6: T.M. M 7y 4 3y 2 X  Soldberg and Case 6: C.R. M 10y1m 4 2y 2 X  Case 5: T.M. M 12y6m 3 2y 2 X  Soldberg and Case 6: C.R. M 10y1m 4 2y 2 X  Case 5: T.M. M 12y6m 3 2y 2 X  Soldberg and Case 6: C.R. M 11y 4 NA 2 X  Case 1: M.F 7y 4 4 4y 2 X  Case 1: M.F 7y 4 4 4y 2 X  Case 1: M.F 7y 4 4 4y 2 X  Case 1: M.F M 12y6m 3 NA 2 X  Soldberg and Case 6: C.R. M 11y 4 NA 2 X  Case 1: M.F M 12y6m 3 NA 2 X  Soldberg and Case 6: C.R. M 11y 4 NA 2 X  Case 1: M.F M 12y6m 3 NA 2 X  Soldberg and Case 6: C.R. M 12y4m 3 2y 2 X  Soldberg and Case 6: C.R. M 12y4m 3 2y 2 X  Soldberg and Case 7: L.N. M 5y2m 4 4y 2 X  Case 1: M.F M 1y 4 NA 2 X  Soldberg and Case 8: M.F M 7y 4 4 4y 2 X  Case 8: M. M 11y 4 NA 2 X  Soldberg and Case 2: R. M NA 4 4y 2 X  Case 2: R. M NA 4 4y 2 X  Case 2: R. M NA 4 4y 2 X  Case 2: R. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X  Case 4: B. M NA 4 4y 2 X		Case 3: B.D.	M	5y	3	5у	1	X			
Case 2:	Cobrinik, 1974		M	12y6m	4	2y	1	X			
Case 3: M   14y   4   Before 5y   2   X		Case 2:	M	14y6m	4	Before 5y	2	X			
Case 4: John   M		Case 3:	M	14y	4	Before 5y	2	X			
Case 6:    M		Case 4: John	M	15y	4	Before 5y	1	X			
Case 6: William W.   14y		Case 5:	M	12y	4	Before 5y	2	X			
Needleman (1976)   Aram et al. (1984)   Case: MD   M   39y   4   4y5m   2   X		Case 6:	M	14y	4	Before 5y	3	X			
Columb   Case   1: M.H.   F   17y8m   4   3y   2   X   X   X   X   X   X   X   X   X	Needleman	Case 1: V.	F	5y8m	3	1y3m	1		X		
Rothermel   Case 2: D.M.   M   6y8m   4   3y   2   X   X				-		-		X			
(1984) Case 3: C.K. M											
Case 4: D.S. M				-							
Case 5: T.M.   M   12y6m   3   4y6m   2   X				-		•					
Case 6: C.R.   M   10y1m   4   2y   2   X   X   X   X   X   X   X   X   X						•		v	X		
Rothermel (1984)         Case 8: A.C.         M         5y2m         4         3y         2         X           (continued)         Case 8: A.C.         M         12y4m         3         2y         2         X           Siegel (1984)         Case: A.E.         F         7y9m         3         3y         1         X           Burd et al. (1985)         Case: B.F.         M         7y         4         4y         2         X           Burd and Kerbeshian (1985)         Case: W.         M         11y         4         NA         2         X           Cossu Marshall (1986)         Case 2         F         18y6m         3         NA         2         X           Burd et al. (1987)         Case 1: A.         F         NA         4         Before 3y         1         X           Case 2: R.         M         NA         4         4y         2         X           Burd et al. (1987)         Case 1: A.         F         NA         4         Before 3y         1         X           Case 2: R.         M         NA         4         4y         2         X           Case 2: R.         M         28y         4 <t< td=""><td></td><td>Case 5: T.M.</td><td>M</td><td>12y6m</td><td>3</td><td>4y6m</td><td>2</td><td>X</td><td></td><td></td><td></td></t<>		Case 5: T.M.	M	12y6m	3	4y6m	2	X			
Rothermel (1984)         Case 8: A.C.         M         5y2m         4         3y         2         X           (continued)         Case 8: A.C.         M         12y4m         3         2y         2         X           Siegel (1984)         Case: A.E.         F         7y9m         3         3y         1         X           Burd et al. (1985)         Case: B.F.         M         7y         4         4y         2         X           Burd and Kerbeshian (1985)         Case: W.         M         11y         4         NA         2         X           Cossu Marshall (1985)         Case 1         F         12y5m         3         NA         2         X           Cossu Marshall (1987)         Case 2.         F         18y6m         3         NA         2         X           Burd et al. (1987)         Case 1: A.         F         NA         4         Before 3y         1         X           Case 2: R.         M         NA         4         4y         2         X           Case 2: R.         M         NA         4         4y         2         X           Case 3: E.         M         28y         4         5y	Goldberg and	Case 6: C.R.	M	10y1m	4	2y	2		X		
(1984) (continued)       Case 8: A.C.       M       12y4m       3       2y       2       X         Siegel (1984)       Case: A.E.       F       7y9m       3       3y       1       X         Burd et al. (1985)       Case: B.F.       M       7y       4       4y       2       X         Burd and Kerbeshian (1985)       Case: W.       M       11y       4       NA       2       X         Cossu Marshall (1986)       Case 1       F       12y5m       3       NA       2       X         Burd et al. (1987)       Case 2       F       18y6m       3       NA       2       X         Burd et al. (1987)       Case 1: A.       F       NA       4       Before 3y       1       X         Case 2: R.       M       NA       4       4y       2       X         Case 3: E.       M       28y       4       5y       2       X         Case 4: B.       M       NA       4       3y       2       X	(1984)	Case 7: L.N.									
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